

# Adenomatoid odontogenic tumor associated with an unerupted mandibular lateral incisor: a case report

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**Abstract** (J Korean Assoc Oral Maxillofac Surg 2015;41:342-345)

Adenomatoid odontogenic tumor (AOT) is a rare, benign odontogenic tumor that predominantly appears in the second decade of life in female patients. Most AOTs occur in the anterior part of the maxilla and are usually associated with impacted anterior teeth. There are three types of AOT, follicular, extrafollicular, and peripheral, which are classified based on the location of the lesion and its association with the impacted tooth. We report a rare case of AOT associated with an impacted right mandibular lateral incisor in an 11-year-old female patient.

**Key words:** Adenomatoid odontogenic tumor, Follicular, Impacted lateral incisor, Mandible

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## I. Introduction

Adenomatoid odontogenic tumor (AOT) was first described by Steensland in 1905. In 1907, Dreiblادت called this lesion a pseudo-adenameloblastoma<sup>1</sup>. Adenameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum, and teratomatous odontoma have also been used to describe the lesions that we currently know as AOT. It was not until 1948 that Stafne considered the lesion as a distinct entity<sup>2</sup>, and Philipsen and Birn<sup>3</sup> proposed the name “adenomatoid odontogenic tumor”. In 1971, the World Health Organization (WHO) adopted Philipsen and Birn’s term, and the WHO currently defines AOT as composed of odontogenic epithelium in various histoarchitectural patterns, embedded in mature connective tissue stroma, and characterized by slow but progressive growth<sup>4</sup>.

AOT is a rare, non-invasive, benign (hamartomatous) epithelial lesion of odontogenic origin and accounts for 2% to 7% of all odontogenic tumors. AOT usually affects young patients, mostly during their second to third decades of life. Women are affected more frequently than men (male:female ratio=1:1.9), and the lesions tend to occur in the anterior maxillary region<sup>5</sup>. There are three types of AOT: a follicular type (73% of all cases), which has a central lesion associated with an impacted tooth; an extrafollicular type (24% of all cases), which has no relation with an impacted tooth; and a peripheral type (3% of all cases)<sup>6</sup>. Of the follicular types of AOT, 36% occur in the mandible. In one study of AOT affecting the mandible, only 28% of cases involved the mandibular incisor area<sup>7</sup>. Here, we present a case of AOT involving the mandible with an unerupted lateral incisor.

## II. Case Report

An 11-year-old female patient visited the Department of Oral and Maxillofacial Surgery of Dankook University Dental Hospital (Cheonan, Korea) with the chief complaint of a two-year history of swelling in the mandibular anterior region. There was mild swelling in the chin area, which was non-tender and had a firm consistency with ambiguous margins.

Intraorally, the patient had distinct swelling in the anterior

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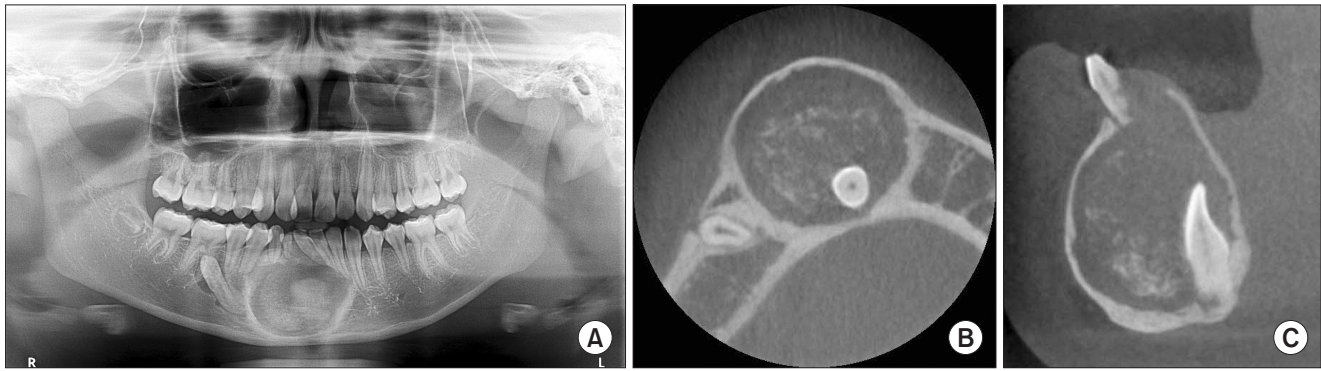
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**Fig. 1.** A. Panoramic view shows lesion of mandible anterior region and impacted mandibular right canine. B. Axial view of computed tomography (CT) scan reveals lesion containing mandibular right lateral incisor and radiopaque foci. C. Sagittal view of CT scan reveals lesion containing mandibular right lateral incisor and radiopaque foci.

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region of the mandible from the left to the right mandibular canines. The overlying mucosa was normal, and there was no paresthesia in the mandibular region. Because of the swelling, the central and lateral mandibular incisors were deviated to the left and the patient had retained her right deciduous lateral mandibular incisor and canine. A panoramic radiograph and computed tomography image were taken, which revealed a well-defined unilocular radiolucency of the anterior mandibular region, which contained an impacted right lateral mandibular incisor with radiopaque foci and an impacted right mandibular canine. (Fig. 1) The differential diagnosis based on the clinical and radiographic findings included a dentigerous cyst, unicystic ameloblastoma, and AOT.

Intraorally, a mucoperiosteal flap was elevated to expose the lesion. The lesion was carefully separated from the surrounding bone, and the lesion and impacted lateral incisor were enucleated. Intraoperatively, the specimen exhibited a solid property and sand-like texture inside. The impacted right mandibular canine was also extracted, but the deciduous teeth were not removed in order to maintain space between the left central mandibular incisor and the right first premolar. The specimen was then sent to the department of pathology for histopathological observation.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

### III. Discussion

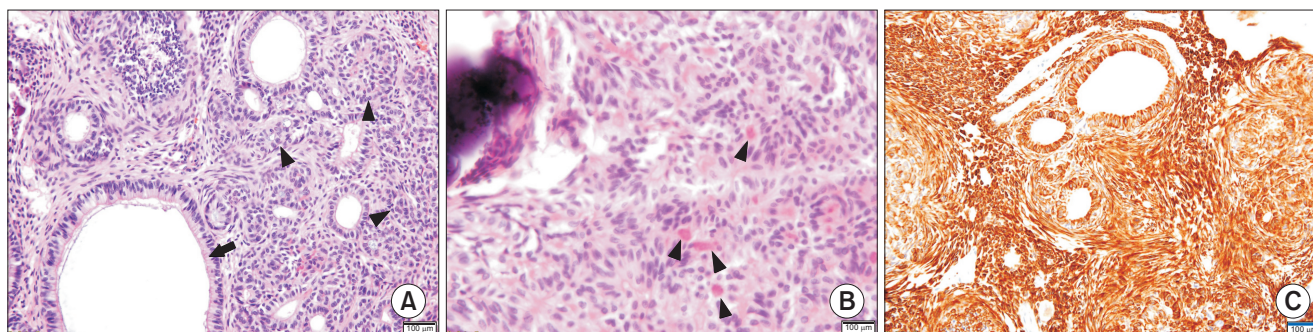
In a review of 272 AOTs by Becker et al.<sup>8</sup>, the patients' ages at the time of diagnosis ranged from 3 to 82 years (mean, 18.4 years). The maxilla-to-mandible ratio was 1.7:1. In 77% of the lesions, small opacities were present, and most were

associated with expansion of the cortical bone. Significant radiological features in patients aged 30 years and older were root resorption and lesions that crossed the midline<sup>8</sup>.

As mentioned above, there are three pathologic types of AOT, intraosseous follicular, intraosseous extrafollicular, and peripheral, all of which have the same histological identity. The follicular type is a central intraosseous lesion associated with an impacted tooth, whereas intraosseous extrafollicular AOT is similar to the follicular type but has no relation with an unerupted tooth. It usually develops around or is superimposed onto adjacent teeth. The peripheral type usually looks like a gingival fibroma or epulis<sup>9</sup>.

Radiographically, AOT is usually unilocular, although a few multilocular cases have been reported. In addition to AOT, the differential diagnosis should include a dentigerous cyst. Radiographically, the pericoronal radiolucency of a dentigerous cyst occurs most frequently in the jaws, and does not extend over the cement-enamel-junction of the tooth. However, an AOT often envelops the crown as well as the root past the cemento-enamel-junction, which distinguishes AOTs from dentigerous cysts. AOTs have numerous, variable-shaped radiopaque foci, which also distinguish them from dentigerous cysts; 78% of AOTs have these foci. Tumor expansion causes displacement of the adjacent teeth, and tooth displacement is more common than root resorption. Irregular root resorption is rare<sup>10</sup>.

This case describes an 11-year-old female patient with pain associated with one of her lateral mandibular incisors. Cortical expansion and dislocation of the adjacent tooth were also present, and there was no sign of root resorption. In cone beam computed tomography, the lesion extended slightly over the cemento-enamel-junction of the impacted tooth and



**Fig. 2.** A. Duct-like structure (arrow) and solid nodule of epithelial cells (arrowheads) (H&E staining,  $\times 100$ ). B. Tumor droplet (arrowheads) (H&E staining,  $\times 400$ ). C. Pan-CK staining,  $\times 200$ .

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contained radiopaque foci. Judging from the association of the impacted tooth, we concluded that this was an intraosseous follicular AOT.

According to the WHO, the histological properties of AOT are as follows<sup>11</sup>: “A tumor of odontogenic epithelium with duct-like structures and varying degrees of inductive changes in the connective tissue. The tumor may be partly cystic and in some cases, the solid lesion may present as a mass in the wall of a large cyst. It is generally believed that the lesion is not a neoplasm.”

Histologically, AOT originates from the odontogenic epithelium and exhibits a solely solid growth pattern or a mixed proportion of solid and cribriform patterns. The histology all AOT types is identical and shows remarkable consistency. The most visible pattern is various solid nodules of columnar or cuboidal epithelial cells that form a rosette-like configuration in the center at low magnification. Most AOTs contain structures with a tubular or duct-like appearance that consist of convoluted structures of epithelium with areas of ductal patterns mixed with globular masses of calcified material<sup>12</sup>. Eosinophilic, amorphous, and uncalcified material (“tumor droplets” or “tumor deposits”) can be found. Philipsen and Reichart<sup>13</sup> and Bravo et al.<sup>14</sup> showed that the amyloid-like eosinophilic deposits represent electron-dense plaques or some form of enamel matrix.

In this case, we found a solid nodule of cuboidal cells and duct-like structures at low magnification (Fig. 2. A) and a tumor droplet at high magnification.(Fig. 2. B)

The AOT phenotype is characterized by cytokeratin staining immunohistochemically. According to Larsson et al.<sup>15</sup>, AOT shows positive staining for CK5, CK17, and CK19, and shows negative staining for CK4, CK10, CK13, and CK18. Crivelini et al.<sup>16</sup> found CK14 in AOT and suggested that its origin was from reduced dental epithelium. In this case, we

conducted Pan-CK staining.(Fig. 2. C) The stimulus that triggers proliferation of AOT is still unknown, although recent studies indicate a histological similarity of AOT to the dental lamina and reduced enamel epithelium. Its resemblance to dental laminae or enamel epithelium remnants implies that AOT is odontogenic in origin. It is usually a hamartomatous lesion rather than a neoplasm<sup>17</sup>.

In conclusion, all types of AOT show encapsulation and benign behavior. The treatment of choice is conservative surgical enucleation, with a very low recurrence rate<sup>9</sup>. In the present case, the lesion and impacted lateral incisor were surgically removed and the patient was without recurrence over the six months of follow-up.

## Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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